CASE REPORT

Iatrogenic Epidermal Inclusion Cyst of the Parapharyngeal Space: Unusual Complication of Ear Surgery

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ABSTRACT

A 46-year-old man presented with a 12-month history of a slow-growing mass at the right anterior temporal and superior parotid region. He had a history of chronic otitis media and had undergone a modified radical mastoidectomy for cholesteatoma 5 years earlier. Physical examination revealed a sinus tract and diffuse soft tissue mass measuring 4 cm in diameter spread throughout the region of the right anterior temporal and superior parotid areas. Magnetic resonance imaging (MRI) showed three separate masses, including contrast material in the right superior parotid region and lateral skull base. The patient underwent a preauricular infratemporal approach. Six months later, a sinus tract recurred at the inferior border of the right zygomatic arch. MRI showed multiple masses in the right prestyloid parapharyngeal space, which were resected through a transparotid approach. The histopathologic diagnosis was an epidermal inclusion cyst (EIC). One year after the operation the patient was in good health and there was no sign of disease.

EICs are rare tumors that are seen when epidermal elements are included in the dermis, which can follow trauma. EICs are unusual in the parapharyngeal space. Thus, until they become clinically observable, primary benign growths may not be recognized in this region. EICs must be considered in the differential diagnosis of growths in the parapharyngeal space, particularly among patients with a prior history of tympanomastoid surgery on the tumor side.

KEYWORDS: Epidermal inclusion cyst, parapharyngeal space, treatment

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An epidermal inclusion cyst (EIC) can be described as a dermal cystic enclosure of keratinizing squamous epithelium that is filled with keratin debris.[1] The terms *traumatic epidermoid cysts* and *EIC* describe the same phenomenon, and both indicate a traumatic etiology.[2] The cause is believed to be the introduction of epidermal elements into the dermis during trauma.[3]

Tympanomastoid surgery, the insertion of ventilation tubes, and stapedectomy are examples of otological procedures that have been reported as etiologic factors in the development of EICs in areas such as the tympanic membrane, middle ear, and temporal fossa.[4–6] A patient with an EIC involving the parapharyngeal space after ipsilateral radical mastoidectomy for cholesteatoma is presented.

ILLUSTRATIVE CASE

A 46-year-old man presented with a 12-month history of a slow-growing mass in the right anterior temporal and superior parotid region. The patient had a history of chronic otitis media and had undergone a modified radical mastoidectomy for cholesteatoma 5 years earlier. His postoperative course was routine. During follow-up, the mastoid cavity appeared well epithelized.

At this presentation physical examination showed a sinus tract and a diffuse soft tissue mass measuring 4 cm in diameter spread throughout the right anterior temporal and superior parotid areas. No lymph nodes on the ipsilateral and contralateral side of the neck were found. Neurosensory functions were normal. A full blood count, and erythrocyte sedimentation rate were within normal parameters. Chest radiography showed no abnormalities.

Magnetic resonance imaging (MRI) showed three separate masses, measuring 3 × 3 cm, 0.5 × 0.5 cm, and 1.2 × 1.5 cm in diameter. The masses were in the right superior parotid region and lateral skull base (Fig. 1). Computed tomography (CT) showed bone erosion of the right lateral skull base (Fig. 2). The patient underwent a preauricular infratemporal approach. During the procedure, osteomyelitis was noted in the sphenoid bone, in particular in the region of the foramen rotundum. The neurosurgical team performed a subtemporal craniotomy. The lesion also affected the dura. The area was resected and the temporal muscle fascia was used for duraplasty

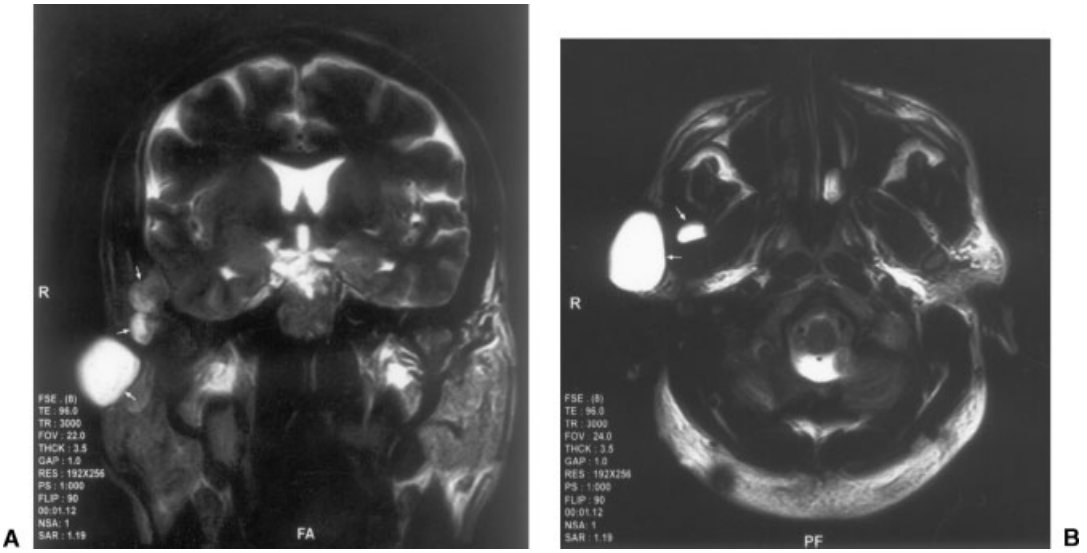

Figure 1 (A) Coronal and (B) axial magnetic resonance images show three separate masses enhanced by contrast material in the right superior parotid region and lateral skull base.

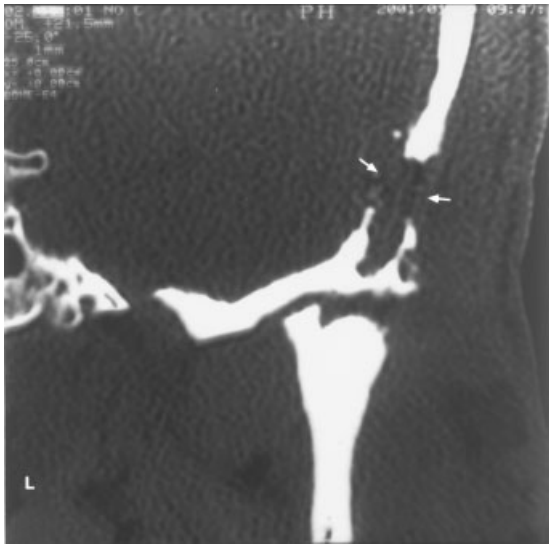

Figure 2 Coronal computed tomograhy shows bone erosion of the right lateral skull base.

(Fig. 3). Phenytoin sodium (300 mg/day) was given to the patient for seizure prophylaxis, and the patient was advised to use it for 8 months. The patient's postoperative course was uneventful. Histopathologic examination of the mass revealed it to be a cholesteatoma, and surgical margins of the dura, which were resected, were reported as clean.

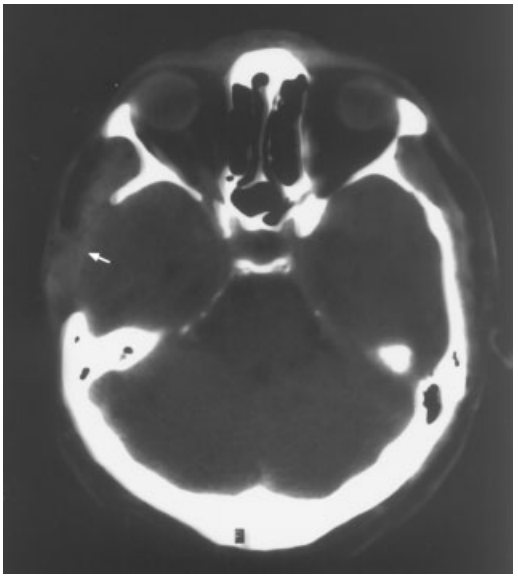

Figure 3 Axial computed tomographic scan after first operation.

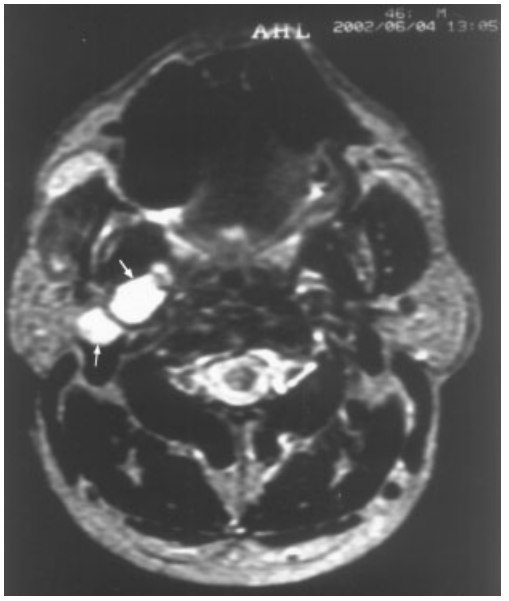

Figure 4 Six-month follow-up magnetic resonance imaging shows multiple masses and contrast material in the right prestyloid parapharyngeal space.

Six months after the operation, a sinus tract recurred at the inferior border of the right zygomatic arch. MRI showed multiple masses with contrast material in the right prestyloid parapharyngeal space. The masses were extremely near the deep lobe of the parotid gland (Fig. 4). The masses were resected through the transparotid approach and submitted for histopathologic analysis.

Histopathologic examination revealed a cystic cavity containing laminated keratin strands. The cavity was lined by keratinized stratified squamous epithelium, confirming the diagnosis of EIC (Fig. 5). After surgery, the patient exhibited House-Brackmann (HB) grade 3 facial nerve paralysis caused by the nerve dissection. Methylprednisolone (1 mg/kg) was administered, decreased incrementally, and stopped in 20 days. Otherwise, the patient's postoperative course proved uneventful and he was discharged on the seventh day after surgery. One year after the operation the patient was in good health and there was no sign of disease. His facial nerve function was evaluated as HB grade 1.

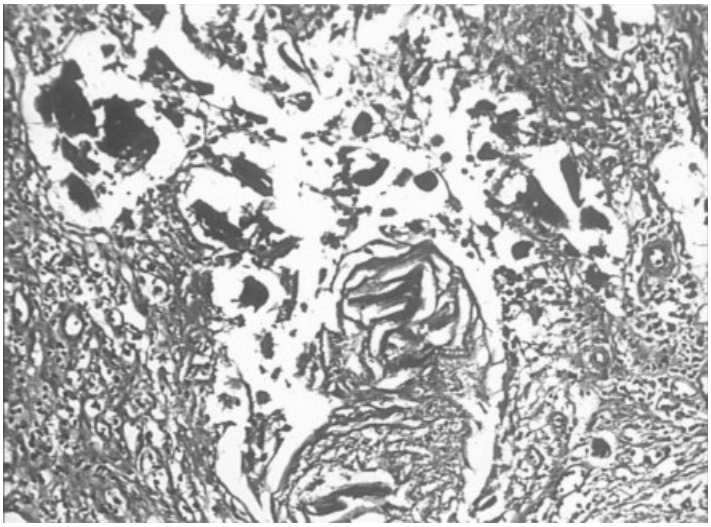

Figure 5 The cavity was filled with keratin (hematoxylin and eosin, ×20).

DISCUSSION

EICs are rare tumors that are seen when epidermal elements are included in the dermis. This situation develops after trauma. The phenomenon is mainly observed in the fingers, palms, and soles.[3] However, EICs also occur in other areas and in tissues deeper than the dermis. The latter has occurred after biopsy procedures or surgery.[2,7,8] EICs usually exist as a single cyst, but more than one may be present.[9]

The epithelium implant theory is the most commonly accepted pathogenesis. It proposes that epidermal structures are driven into deeper tissues. The trapped epidermal structures assume the role of a skin graft and become independent. The tissue continues growing in its new position and produces keratin, thus forming a cyst.[10]

Clinically, these cysts manifest as painless, slow-growing, well-circumscribed swellings and may occur at any time from adolescence to adult life.[10] EICs are invariably small but occasionally reach a diameter of 5 cm.[11] When a cyst is present, the symptoms vary depending on where it is located in conjunction with the amount of pressure it exerts on surrounding structures.[3] As a rule, epidermal cysts are slow growing. However, an increased amount of desquamated epithelium or a reaction in the surrounding soft tissue may lead to a sudden enlargement.[12] Symptoms may appear as soon as 6 months after injury or may not appear until 20 years later.[10,13]

EICs lack ectodermal appendages, such as hair, nerve, bone, teeth, and sweat glands, that are associated with dermoid cysts.[14] Defined as a tissue, the cyst wall is composed of stratified epithelium and lumen that contain keratin in addition to desquamated cells, which are found in laminated layers. Occasionally, the walls may be calcified.[12] If the cyst ruptures, the result may be a granulomatous foreign body reaction, which can be diagnosed erroneously as an infection of the cyst.[2]

As a result of ipsilateral radical mastoidectomy, EICs have been found in the upper neck region.[9] Similarly, the following may lead to the formation of cysts: harvest of temporal fascia in the infratemporal fossa or parotid gland and facial nerve decompression in the infratemporal fossa.[4,12] EICs may form after these procedures, possibly because the disease moves through the mastoid area into these adjoining regions by direct propagation.[9]

Studies show that EICs are unusual in the parapharyngeal space. Thus, until they become clinically observable, primary benign growths may not be recognized in this region.[2]

In our patient, the hisopathologic diagnosis of an EIC was likely a secondary complication of his surgery for chronic otitis media, which had been

performed 5 years previously. Pertinent literature seems to support our diagnosis.

For treatment, these cysts must be excised completely, including any attached skin containing the pore. Incomplete excision leads to recurrence of the cyst.[3]

After ear surgery, all otolarygologists must be aware of the possibility of EICs developing in adjacent areas.[9] EICs must be considered in the differential diagnosis of growths in the parapharyngeal space, particularly in patients with a prior history of tympanomastoid surgery on the tumor side.[4]

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Commentary

The authors report a "complication" associated with a prior otolaryngological procedure for chronic otitis media. They review the findings associated with an epidermal inclusion cyst and detail the associated pathology. Given the intracranial extension of this process, they comment on the neurosurgical team's involvement in performing a subtemporal approach.

Epidermoid cysts resulting from prior procedures are relatively well known. However, it is rare to find a case with the qualities of this particular report; that is, a recurrent intracranial epidermal inclusion cyst requiring combined treatment with neurosurgery.

This report alerts otolaryngologists to possible complications associated with this surgical procedure and cautions that it may be advantageous to use the neurosurgical team to approach the intracranial aspects of the cyst and to ensure a meticulous closing to avoid leakage of cerebrospinal fluid.

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